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Case Report

Long term outcome of tibial adamantinoma: a case report and literature review

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ABSTRACT

Adamantinoma is a rare malignant bone tumor that typically occurs in the tibia but can also occur in the jaw, forearm, hands, or feet. It commonly affects young adults between 20 and 40 years. It accounts for less than 1% of primary bone tumors. It is a slow growing tumor and often indolent but generally symptoms are persistent pain in the affected area, localized swelling, tibial bowing deformity and pathological fracture which occurs acutely. the diagnosis is based on imaging and a necessary biopsy to confirm the diagnosis, showing nests of epithelial-like cells in a fibrous stroma. The treatment consists of surgical resection and recurrence can be problematic and may occur even decades after initial treatment. Here, we report one case of tibial adamantinoma with atypical treatment and long-term evolution greater than 24 years and with no recurrence.

Keywords: Adamantinoma, Diagnosis, Review, Treatment

INTRODUCTION

Adamantinoma is a rare, malignant bone tumor primarily found in the tibia (shin bone). Characterized by its slow growth and locally aggressive behaviour, this tumor affects young adults, more commonly men, typically between 20 and 40 years old. Despite its rarity, adamantinoma has a potential to cause significant morbidity and it is characterized by a unique histological appearance, resembling epithelial cells amidst a fibrous stroma. The primary treatment for adamantinoma is wide-margin surgical resection, aiming to ensure complete removal and prevent recurrence.[1-7]

METHODS

32-year-old patient hospitalized for pathological fracture of the tibia occurred during a football match. An X-ray of the leg showed a fracture in the middle of the diaphysis with a normal appearance of the upper part of the diaphysis and a lower part under the fracture showing abnormal lesions as multiloculated osteolytic lesions with a thin sclerotic rim. The patient reported that in his medical history he did not present significant pain, nor swelling, nor inflammatory signs, nor any apparent particular impotence. Given this radiological aspect, the first question was to know to what these lesions corresponded in order to have an idea of the diagnosis and what treatment to plan. After consultation with different colleagues, three diagnoses were retained, fibrous dysplasia, chronic tuberculous osteomyelitis, and adamantinoma. The only way to manage treatment was to do a biopsy and temporarily fix the fracture until the diagnosis was confirmed. We proceeded to a careful intramedullary curettage of all the bone and specially the diseased diaphysis with the same technique for chronic

osteomyelitis, our thoughts about diagnosis were, before biopsy result, a tuberculous osteomyelitis and we immobilized the tibia with an Ender nail with temporarily intention. Unfortunately, after the operation, the patient, who is from rural origin, was lost to follow-up for almost two months, in the meantime the diagnosis of amandatinoma was made. After several phone contacts, the patient returned for consultation and the radiological control was rather reassuring, the bone was in the process of consolidation and the lesions showed significant signs of healing. We decided to keep the same course of action and to monitor patient's progress. After a one-year follow-up, the fracture was completely consolidated, and the soap bubble lesions had greatly regressed. After 3 years, we carried out an ablation of the nail and we took the opportunity to repeat the biopsy which histological result showed inflammatory type lesions. Now the patient arrived to 24 years follow up and he has not suffered any recurrence and without neither clinical-signs but with perseverance of slightly lacunar appearance radiologically on plain X-rays and CT but with a healed appearance in comparison with the first time.

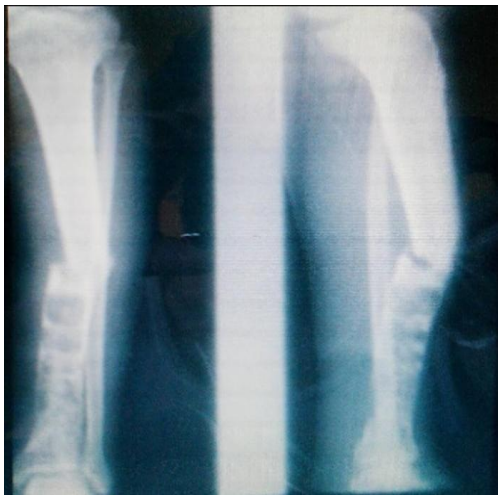

Figure 1: Pathological fracture caused by an adamantinoma.

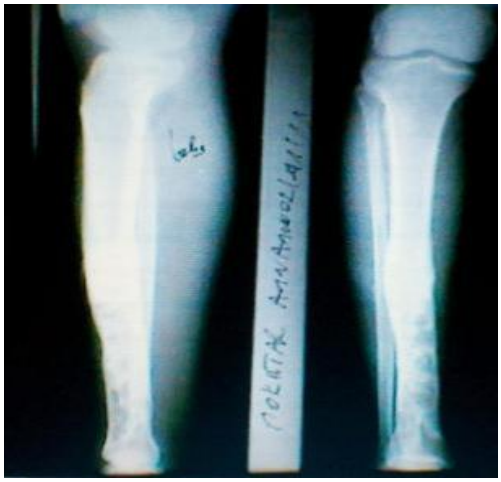

Figure 2: Radiological appearance after 11 years.

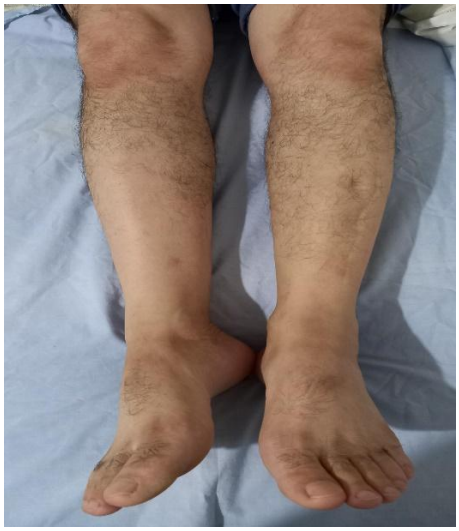

Figure 3: Clinical appearance after 24 years.

Figure 4: CT showing perseverance of slightly lacunar lesions after 24 years.

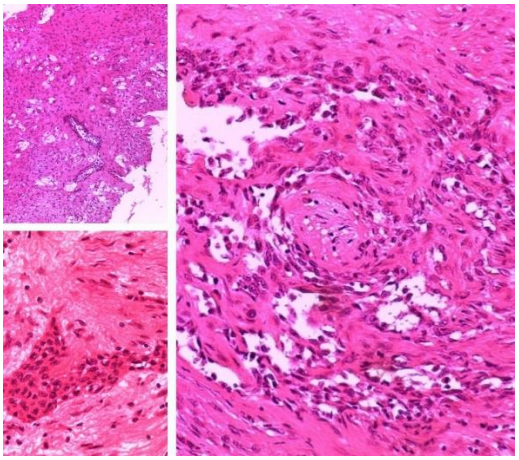

Figure 5: Histological appearance showing epithelial cells amidst a fibrous stroma.

DISCUSSION

Adamantinoma is a rare, slow-growing bone tumor, most commonly affecting the tibia (the shin bone). It accounts for less than 1% of all bone tumors and typically occurs in adults, with a slight male predominance between the ages of 20-35.[1-7] Adamantinoma can also occasionally affect the fibula or other bones but is primarily associated with the long bones of the legs.

Our patient was 32 years old man and was hospitalized for pathological fracture of the tibia with abnormal lesions of the lower half of the tibia as multiloculated osteolytic lesions with a thin sclerotic rim. The fibula was not affected.

Clinically the symptoms are pain and swelling in the affected area, with symptoms that may persist for a long period before diagnosis. It can also cause deformities as bowing of the tibia which can occur or fractures in advanced stages, the tumor can weaken the bone, leading to a fracture.[2,3,5]

In his medical history our patient did not report any pain, swelling, inflammatory signs or impotence before the fracture he got while he was playing football.[4] For diagnosis , imaging as X-rays, MRI, and CT scans are used to identify the tumor and assess its size and location.[1-4] Radiologically, it often shows a well-demarcated, <<soap bubble-like >>appearance and well-circumscribed, expansile , multiloculated lesion with a thin sclerotic rim. MRI is essential for defining the extent of the disease pre-operatively and biopsy for definitive diagnosis (Figure 5).

The patient was hospitalized in 2000 and at that date we did not have an MRI in our hospital structure but the lesions in plain X-rays showed exactly multiloculated osteolytic lesions with a thin sclerotic rim. In our country osseous tuberculosis is quite frequent, so our thoughts among differential diagnosis was that diagnosis before biopsy. Effectively after biopsy, the result was an adamantinoma showing histologically epithelial cells amidst a fibrous stroma (Figures 1,5).

Adamantinoma should be differentiated from chronic osteomyelitis, notably tuberculous osteomyelitis which is frequent in our country, and interfibrous dysplasia, which has similar radiologic characteristics but tends to be less aggressive. Concerning treatment, the primary treatment is surgical removal (wide excision), limb salvage surgery, reconstruction, amputation, curettage. In cases of recurrence or metastasis, more aggressive surgical approaches or therapies might be needed.[4-7]

We proceeded to a careful intramedullary curettage of all the bone and specially the diseased diaphysis with the same technique for chronic osteomyelitis, our thoughts about diagnosis were, before biopsy result, a tuberculous osteomyelitis and we immobilized the tibia with an Ender nail with temporarily intention.

The therapeutic protocol in the management of adamantinoma is not formally established. Braud et al. identified three methods (curettage, resection and amputation) used without asserting the superiority of one or another. P Baud reported that judgement could be not made as the superiority of one technique over another. For additional treatments radiation and chemotherapy are generally not effective for adamantinoma and are not typically used.

About evolution, metastasis, although rare at initial diagnosis, adamantinoma can spread to other parts of the body, such as the lungs, in about 15-20% of cases. For our patient after a one-year follow-up, the fracture was completely consolidated, and the soap bubble lesions had greatly regressed, which means that there has been bone regeneration and regression of the initial lesions. Now the patient arrived to 24 years follow up and he has not suffered any recurrence and without neither clinical sign but with perseverance of slightly lacunar appearance radiologically on plain X-rays and CT but with a healed appearance in comparison with the first time.

About prognosis, survival rate with proper treatment, the prognosis for tibial adamantinoma is generally good, with about 85% of patients surviving for 10 years or more after diagnosis. Patient has now more than 24 years survival.[4,5,7]

CONCLUSION

Adamantinoma is a rare, low-grade malignant bone tumor primarily affecting the tibia. It presents with pain, swelling, and potential deformity, sometimes it is clinically mute. Diagnosis is confirmed through imaging and biopsy. Treatment involves wide surgical excision with reconstruction, curettage even used by some authors and used by us by ignorance of diagnosis before biopsy is not recommended even with 24 year's survival of our patient. Life-long follow-up is essential due to the risk of local recurrence and metastasis. Prognosis is generally favorable with appropriate management.

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